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Taste Testing in a Pediatric Case of Congenital Aglossia

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Abstract

Objective: Congenital aglossia (CA) is a rare inborn condition characterized by absence of the tongue. This study aimed to determine gustatory function in a pediatric female participant with CA. Methods were developed based on previous taste discrimination research of a participant with isolated congenital aglossia (ICA) and anecdotal reports of individuals with CA possessing the ability to discern taste stimuli.

Methods: In this randomized, double-blinded trial, 78 samples were presented to an 11-year-old Hispanic female with CA in the presence of her parents. Triplicate samples of sucrose (sweet), citric acid (sour), sodium chloride (salty), caffeine (bitter) and monosodium glutamate (umami) solutions at five concentration levels each were tested, with three water samples. Contingency tables were developed with participant responses. Descriptive statistics were run to determine accuracy of stimuli identifications, with correct identification set at ≥ 66.6% accuracy (2 out of 3). Taste identification accuracy versus stimuli taste, concentration, and presentation order were tested for statistically significant association (p ≤ 0.05) using Chi-square testing.

Results: Out of five stimuli, the participant correctly identified sour and umami. Umami was accurately identified 100% of the time, and sour 66.6% of the time. Correct identification threshold criteria were not met for sweet, salty, and bitter samples. There was no statistically significant association between taste identification accuracy and particular taste, concentration levels, or presentation order. Bitter taste was commonly recorded as sour, and bitter as salty.

Conclusion: This study supports previous findings indicating that people with CA may accurately identify sour, though at a higher threshold than previously found in the ICA trial or in persons without CA. Compared to the previous trial, the participant in this trial could accurately identify umami. Higher discrimination thresholds of sour and umami, and misidentification of sweet, salty, and bitter stimuli, may indicate gustatory impairment among individuals with CA.

Keywords: Congenital aglossia; Taste; Flavor; Nutrition status; Rehabilitation; Quality of life

Introduction

Congenital aglossia (CA) is a very rare inborn condition characterized by the absence of the tongue. It may present with other developmental malformations such cleft palate, lower lip defects, and limb abnormalities [1]. However, cases of isolated congenital aglossia (ICA) in which a missing tongue is identified as the sole anomaly are also documented. The first report of a patient with CA stems from 1718 when de Jussieu reported on a child born without a tongue [2]. Most of the following research and publications on CA are composed of clinical case reports. Some of these reports provide anecdotal evidence on taste function; however, few include specifics on taste discrimination using systematic tests.

Eskew and Shepard reported on the case of a 22-year-old man who was taste tested at Washington University Dental Clinic as part of the clinical examination of his case [3]. Solutions of 3% cane sugar, 1% sodium chloride, 0.1% sulfuric acid, and 0.1% quinine were brushed on his oral cavity to determine taste perception. The researchers

reported that the participant appeared to detect taste on the surface of his soft palate. However, details on stimuli recognition were not provided [3]. Salles et al. reported on a 14-year-old girl with CA who showed difficulties suckling and swallowing after birth and required a nasogastric tube for a short amount of time to ensure adequate nutrition. She later developed eating mechanisms to consume her family's usual diet [2]. A taste test was conducted with six increasing concentrations of four taste stimuli: sweet, acid, bitter, and salty. The participant was able to correctly identify acid taste at the lowest concentration of 0.015 g/L and salty at the fifth concentration of 1.5 g/L, while bitter and sweet were not correctly identified at any concentration level [2]. Results suggested that the patient showed a greater sensitivity to acid and salty stimuli than to sweet and bitter tastes, though this did not appear to affect her everyday food choices [2].

The current study involving a pediatric case with CA builds on a randomized, double-blinded, controlled taste test trial by McMicken et al. reporting on the taste function of a 44-year-old woman with ICA [4]. Unlike other cases of CA, this individual's sole anatomic anomaly was the absence of the tongue. The study was the first to include umami as one of the stimuli. Five concentration levels of sweet, salty,

sour, bitter, and umami were randomly presented to the participant who was able to accurately identify sweet at 17.1 g/L, salty at 5.8 g/L, sour at 0.033 g/L, and bitter at 0.02 g/L as the lowest concentration thresholds [4]. The participant did not correctly identify umami at any concentration level.

This current study aimed to further investigate the taste function of individuals with CA. As previously shown, while reports on taste function in this population exist, few systematically tested the abilities and limitations of taste recognition.

Methods

This study and its design were based on the taste test trial with a person with ICA by McMicken et al. [4]. Only one participant was recruited for this study, selected based on her diagnosis of CA and her parents' agreement to allow her to participate in the study. The 11-year old Hispanic girl and her mother reported that she consumed her family's usual diet. The California State University, Long Beach Institutional Review Board approved this study's procedure. Parental written consent and participant verbal assent were obtained prior to trial initiation.

Sample solution preparation and data collection were completed in the Food Science laboratory at California State University, Long Beach. Solutions for each of the five taste stimuli were prepared using sucrose (C&H) for sweet, sodium chloride (NaCl; Morton) for salty, caffeine (Sigma Aldrich) for bitter, citric acid (Now Foods) for acidic, and monosodium glutamate (MSG; Ajinomoto) for umami. Five increasing levels of concentration strength were prepared for each taste, yielding 25 unique solutions (Table 1). Taste trial preparation and procedures, as well as concentration levels of sweet, salty, bitter, and umami were based on the taste test trial by McMicken, et al [4]. The ICA trial had included acetic acid as sour stimulus. Since acetic acid has a potential olfactory profile, the participant used a nose clip to avoid any interference of gustatory discrimination by olfactory influence. The participant reported difficulty breathing with the nose clip in place, which resulted in a potentially shorter duration of solution exposure to the oral cavity, and thereby a possibly diminished sensitivity during the taste testing [4]. To avoid this issue in the current trial, acetic acid was replaced by citric acid. Concentration levels of citric acid prepared for this trial were based on normal taste threshold and the taste test trial with a person with CA by Salles, et al. [4,5].

Sweet Sucrose	Salty NaCl	Sour Citric Acid	Bitter Caffeine	Umami MSG
0.5	0.1	0.00032	0.001	0.01
0.05	0.05	0.00016	0.005	0.005
0.005	0.01	0.00008	0.0001	0.001
0.001	0.005	0.00004	0.00005	0.0005
0.0005	0.001	0.00002	0.00001	0.0001

Table 1: Stimuli Solution Concentrations in M.

Sample solutions of sucrose, NaCl, citric acid, and MSG at the different concentration levels were prepared by simple dilution using 1 M stock solutions. Due to difficulties with solubility of the caffeine solute when attempting to prepare a 1 M stock solution, bitter solutions were prepared separately for each concentration level by manually dispersing weighed caffeine powder in solution. Solution preparation took two days and was completed two days prior to the taste test trial. Flasks were sealed and stored in the laboratory refrigerator. A total of 25 unique solutions were prepared to be divided into triplicate samples. An additional three water samples were included, yielding 78 sample units to be taste tested.

A research assistant not involved in solution preparation poured three servings of approximately 60 mL of each solution into 78 sample cups, labeled each cup with a randomized three-digit number, and recorded each cup's number, taste stimulus, and concentration level. The taste testing was conducted by a research assistant blinded to the randomization process previously described. The participant and her mother were pre-briefed and the experimental process was discussed verbally. The participant was instructed to take a sip of the solution, keep the solution in her mouth and swish it around for as long as desired without swallowing the liquid, and then discard it into a spittoon. Additional sips of the solution were permissible if any was left in the cup. The participant was asked to identify the solution and then rinse her mouth with distilled water. Answering choices were limited to "sweet", "salty", "sour", "bitter", "umami", "water", "I can't tell", or selecting multiple taste stimuli. As an additional aide to assist with selection, a simple hand-written list with the different options was provided to the participant at the 17th sample. A 10 to 20 minute break was held after every 20 samples.

Data was analyzed using IBM SPSS Statistics for Windows (Version 22.0). The recorded responses were used to develop contingency tables and descriptive statistics were run to determine percentage of accurate stimuli identification and frequency of substitutions. Correct identification was set at a threshold value of $\geq 66.6\%$ accuracy (2 out of 3). A heat map was created to display frequencies of perceived versus actual taste. Cross-tabulations of taste identification accuracy versus actual taste, stimuli concentration, and presentation order were developed and tested for statistically significant association ($p \leq 0.05$) using Chi-square testing.

Results

A total of 78 samples were tasted tested to include five increasing concentration levels of sucrose (sweet), citric acid (sour), NaCl (salty), caffeine (bitter), and MSG (umami), as well as three blank water samples. Out of the 78 samples, only 16 were correctly identified as their actual stimulus or water. Three samples were identified as their actual taste in combination with an incorrect identification: bitter and umami for a bitter solution, sour and bitter for a sour solution, and salty and sour for a salty solution. Using a threshold value of $\geq 66.6\%$ (2 out of 3) for correct identification, only sour and umami were correctly identified at one concentration level each (Table 2).

NaCl (Salty)		Citric Acid (Sour)	
M	**Identified**	**M**	**Identified**
0.1	33.30%	0.00032	33.30%
0.05	0%	0.00016	66.60%
0.01	33.30%	0.00008	0%
0.005	0%	0.00004	33.30%
0.001	0%	0.00002	33.30%
Sucrose (Sweet)		MSG (Umami)	

M	Identified	M	Identified
0.5	33.30%	0.01	33.30%
0.05	0%	0.005	100%
0.005	33.30%	0.001	33.30%
0.001	33.30%	0.0005	0%
0.0005	0%	0.0001	0%
*Caffeine (bitter) was accurately identified zero percent of the time at any concentration level			

Table 2: Percent Samples Accurately Identified by Concentration*.

Actual Taste	% Realized	Combination
Bitter	0%	Bitter + Umami
Salty	13.30%	Salty + Sour
Sour	33.30%	Sour + Bitter
Sweet	20%	
Umami	33.30%	
Water	33.30%	

Table 3: Percent Samples Accurately Identified by Taste.

Sour solution was correctly identified 66.6% of the time at the second strongest concentration level of 1.6×10^{-4} M, 33.3% at 3.2×10^{-4} M and 4×10^{-5} M, 0% at 8×10^{-5} M, and 33.3% at 2×10^{-5} M. Umami solution was correctly identified 100% at the time at the second strongest concentration level of 5×10^{-3} M, 33.3% at 0.01 M and 1×10^{-3} M, and 0% at 5×10^{-4} M and 1×10^{-4} M. Sweet was correctly identified 33.3% of the time at 0.5 M, 5×10^{-3} M, and 1×10^{-3} M, but 0% of the time at levels of 0.05 M and 5×10^{-4} M. Salty was correctly identified 33.3% of the time at 0.1 M and 0.01 M, but 0% of the time at 0.05 M, 5×10^{-3} M, and 1×10^{-3} M. Bitter was correctly identified 0% of the time at any concentration level.

The most common taste substitution was bitter identified as sour at five times, followed by bitter identified as salty at four times (Figure 2). For other taste stimuli, the most common taste substitutions were mistaking sweet as sour or salty at three times each, sour as water (n=3), salty as sour (n=3), and umami as water (n=3).

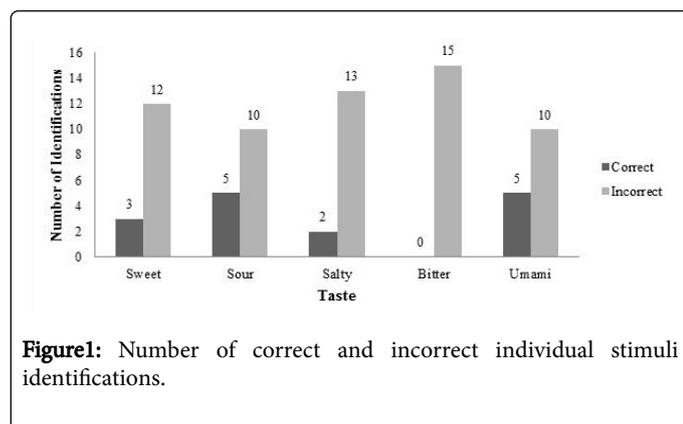
Figure 2: Frequency of Taste Substitutions.

Figure1: Number of correct and incorrect individual stimuli identifications.

In terms of individual sample identification, out of 15 samples per stimulus, the participant most frequently identified both sour and umami taste correctly at five total accurate identifications each, followed by sweet at three correct identifications, and then salty at two correct identifications (Figure 1). Bitter was not correctly identified as the sole stimulus at any attempt. Overall, out of all samples presented, both sour and umami were correctly identified 33.3% of the time, sweet 20% and salty 13.3% of the time (Table 3). Sour and umami taste both had the highest amounts of correct individual sample identifications while also meeting the threshold for accurate taste identification.

A statistically significant association (p ≤ 0.05) could not be determined between taste identification accuracy and particular taste [$X^2(5)$=7.674, p=0.175] or individual stimuli concentration levels (sweet p=0.645; sour p=0.558; salty p=0.484; umami p=0.061). The order of presentation of samples was recorded starting with the 21st sample presented. No statistically significant association was determined between taste identification accuracy and presentation order [$X^2(5)$=55.000, p=0.437].

Discussion

In this taste trial with an 11-year old girl with CA, the participant correctly identified sour and umami taste samples, while sweet, salty, and bitter were inaccurately identified. In the taste test trial with a person with ICA by McMicken et al., the lowest concentration strength the participant was able to correctly identify using the same accuracy threshold of 66.6% (2 out of 3) was sweet (sucrose) at 0.05 M, salty (NaCl) at 0.1 M, sour (acetic acid) at 5×10^{-4} M, and bitter (caffeine) at 1×10^{-4} M [4]. The participant incorrectly identified umami (MSG) at all concentration levels. Compared to the ICA trial, this 11-year old participant with CA was able to correctly identify the umami stimulus at the second highest concentration of 5×10^{-3} M. This is a new finding, which was not established in any of the previous studies looking at taste function in patients with CA. Compared to the reported normal population threshold for MSG at 5×10^{-4} M, the participant did require a higher stimulant concentration for correct identification [5].

In this trial, sour was correctly identified at the second highest concentration of 1.6×10^{-4} M. This threshold is higher than the threshold findings by Salles et al. of 7.8×10^{-5} M and the normal taste threshold of 7×10^{-5} M [2,5]. While sour was one of the tastes correctly identified in the trial by McMicken et al., a direct comparison regarding concentration threshold cannot be made as a different stimulus, acetic acid, was used.

Interestingly, both umami and sour were correctly identified at the second highest concentration level presented, while only achieving 33.3% accuracy at the highest concentration levels. This may suggest a narrower window of taste identification range or sensory overload at

higher concentrations. Further research is needed to explore possible impairments in detecting solutions at higher concentrations.

Unlike in the taste test trials by Goto et al., Salles et al., and McMicken et al., this pediatric participant with CA did not correctly identify sweet, salty, or bitter testing solutions [4-6]. Notably, while sweet and salty were identified 33.3% of the time at select concentration levels, bitter was not correctly identified as the sole stimulant at any attempt. The results of this study partially reflect the findings of Salles et al. by suggesting a decreased sensitivity to bitter stimulus and greater sensitivity to sour taste. The person with ICA detected sweet, salty, and sour stimuli at higher concentrations than had been reported in the normal population, while bitter was identified at a lower threshold than in the normal population [4].

The tongue is the primary organ for taste stimuli detection and discrimination. Taste is a crucial part of nutrition and influences food choices [7,8]. It affects food intake, nutritional status, and quality of life [9]. While CA is a very rare condition, secondary causes may result in an individual's partial or total tongue resection. Tongue cancer requires surgical treatment in 5-40% of cases [10]. A meta-analysis of studies reporting on taste function scores of head and neck cancer patients indicated a significant difference in taste ability before and after treatment [11]. Assessment of health-related quality of life survey data in patients after primary surgery for oral tumor resection indicated that taste function was negatively affected after surgery along with chewing, speech, and swallowing function [12]. Head and neck cancer patients undergoing multimodal therapy frequently reported alterations in taste to decrease appetite, desire to eat, and enjoyment of food [13]. Studying taste discrimination in CA subjects may assist in supporting individuals who underwent total or subtotal glossectomy in obtaining optimal nutrition and improve quality of life.

This study's results are presented with several limitations in mind, including that the participant was an 11-year-old child. Since taste testing was conducted in a research laboratory in the presence of her parents, the surroundings and presence of researchers and family may have influenced her ability to allow for accurate taste discrimination. Perceived pressure to perform and provide assumed correct answers may have influenced the participant despite procedure instruction prior to the test. Further limitations include the participant's medical history briefly reported by the parents, which included recent oral surgery that may have affected her taste sensitivity.

Ultimately, in addition to the results of previous taste test trials, this study's findings of higher discrimination thresholds for sour and umami taste compared to normal taste thresholds, as well as the misidentification of the sweet, salty, and bitter stimuli may indicate possible gustatory impairment among some individuals with CA compared to persons born with a tongue. However, gustatory function in this population may ultimately be highly individualized based on co-occurring anomalies. Further research is needed to determine these factors and gather more data for comparison. Due to the extreme rarity of this condition, this study provides one more trial involving a participant with CA. Future studies may be able to provide additional data on taste thresholds and discrimination function among other persons with CA or loss of tongue due to secondary causes.

Acknowledgement

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